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## The developmental trajectory of parent-report and objective sleep profiles in Autism Spectrum Disorder: Associations with anxiety and bedtime routines

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| Abstract:        | <p>The present study compared the course of parent-report and actigraphy derived sleep profiles over a one-year period, in school-age children with Autism Spectrum Disorder (ASD) and typically developing (TD) children. The Children's Sleep Habits Questionnaire and fourteen nights of actigraphy were used to assess sleep profiles. Parents also completed the Spence Children's Anxiety Scale, the Social Worries Questionnaire, and the Bedtime Routines Questionnaire. Between-group differences in parent-reported sleep problems were less pronounced at follow up, compared to baseline. The course of objective sleep was comparable between groups, with a significant reduction in sleep duration over time in both groups. Children with ASD were further characterised by significantly more night-to-night variability in sleep quality, across both time points. Reductions over time in parent-reported sleep problems were significantly associated with reduced anxiety. Reductions in actigraphy derived sleep efficiency were associated with an increased frequency of maladaptive activities in the hour before bedtime, in both children with and without ASD.</p> |

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Abstract

The present study compared the course of parent-report and actigraphy derived sleep profiles over a one-year period, in school-age children with Autism Spectrum Disorder (ASD) and typically developing (TD) children. The Children’s Sleep Habits Questionnaire and fourteen nights of actigraphy were used to assess sleep profiles. Parents also completed the Spence Children’s Anxiety Scale, the Social Worries Questionnaire, and the Bedtime Routines Questionnaire. Between-group differences in parent-reported sleep problems were less pronounced at follow up, compared to baseline. The course of objective sleep was comparable between groups, with a significant reduction in sleep duration over time in both groups. Children with ASD were further characterised by significantly more night-to-night variability in sleep quality, across both time points. Reductions over time in parent-reported sleep problems were significantly associated with reduced anxiety. Reductions in actigraphy derived sleep efficiency were associated with an increased frequency of maladaptive activities in the hour before bedtime, in both children with and without ASD.

Keywords

Autism Spectrum Disorder, sleep problems, actigraphy, school-age children, anxiety, bedtime routines

## Introduction

Parents of school-aged children with Autism Spectrum Disorder (ASD) report sleep problems at a consistently higher rate than parents of typically developing (TD) children (Hodge, Carollo, Lewin, Hoffman, & Sweeney, 2014; May, Cornish, Conduit, Rajaratnam, & Rinehart, 2015; Souders et al., 2009). The increased prevalence of parent-reported sleep problems is accompanied by heightened bedtime resistance, sleep onset issues, sleep anxiety and parasomnias (Hodge et al., 2014; May et al., 2015; Souders et al., 2009). Furthermore, the use of objective sleep assessment methods identify a significantly longer sleep onset latency (SOL) in school-aged children with ASD relative to TD children (Allik, Larsson, & Smedje, 2006; Souders et al., 2009). Although the presence of disturbed sleep is well established in school-aged children with ASD, little is known about the continuity and change of these disturbed sleep profiles during this key developmental period.

The school-aged years are characterised by an array of cognitive, physical and social developments, during which time sleep patterns begin to mature in preparation for adolescence (Sheldon, 2014). The development of sleep during this time is relatively well defined in TD children. There is a gradual delay in bedtime and sleep onset times, and a consequent decrease in sleep duration; with this effect being most pronounced from middle childhood onwards (Blair et al., 2012; Kelly & El-Sheikh, 2011; Pesonen et al., 2014; Sadeh, Dahl, Shahar, & Rosenblat-Stein, 2009). Around 40% of TD children are reported to experience sleep problems during this time, and these sleep-problems manifest as chronic conditions in up to 60% of cases (Fricke-Oerkermann et al., 2007; Pesonen et al., 2014). The extent to which these developmental trajectories compare to children with ASD is unclear however, with few longitudinal studies conducted to date. An assessment of parent-report sleep problems over a 12 month period identified a significant reduction in the severity of sleep problems in school-aged children with ASD. Notably, no significant change was seen

over the same period of time in TD children (May et al., 2015). This coincides with recent cross-sectional evidence that the magnitude of parent-report sleep problems in children with ASD may be most pronounced during early childhood (Hodge et al., 2014). Furthermore, Humphreys et al. (2014) compared parent-reported sleep duration and night wakings from birth to 11 years, in children with and without ASD. Children with ASD were characterised by increased night wakings and shorter sleep duration from 30 months onwards. Importantly, the greatest group differences in both night waking and sleep duration were observed between 6-7 years of age (i.e., 81 months). To our knowledge, only one study to date has assessed sleep objectively over time in school-aged children with and without ASD. Allik, Larsson, and Smedje (2008) used actigraphy to compare the trajectory of sleep in children with and without ASD over a 2-3 year period. Overall, the trajectory was similar across groups, with a gradual sleep phase delay, shorter sleep duration and decreased sleep efficiency over time. This suggests that whilst objective sleep profiles mature at a similar rate, the trajectory of parent-report sleep problems in children with ASD may diverge from that of TD children. With such a scarcity of research, the current study aims to assess the short-term trajectory of both subjective and objective sleep profiles in school-aged children with and without ASD.

Capturing the course of sleep profiles over time provides an opportunity to investigate the association between individual changes in sleep and other key factors. The current study focuses on the complex and bi-directional association between sleep and anxiety (Leahy & Gradisar, 2012). Based on the seminal model proposed by Dahl (1996), it is understood that anxiety inhibits sleep via increased hyper-vigilance and arousal. An understanding of the nature of this relationship in children with ASD is of particular importance, given that the prevalence of co-morbid anxiety symptoms is reported to be as high as 84% (White, Oswald, Ollendick, & Scahill, 2009). Mirroring previous findings in TD children (Alfano, Zakem,

Costa, Taylor, & Weems, 2009; Fletcher et al., in press; Gregory & Eley, 2005), there is a well-established concurrent association between parent-reported sleep problems and anxiety symptoms in children with ASD (Goldman et al., 2011; Hollway, Aman, & Butter, 2013; Mayes & Calhoun, 2009; Rzepecka, McKenzie, McClure, & Murphy, 2011). Most recently, May et al. (2015) assessed sleep and anxiety over a one year period and found that parent-reported sleep problems predicted later anxiety in school-aged children with ASD. Although children with ASD had significantly higher levels of anxiety, the relationship was not mediated by ASD diagnosis (i.e., the relationship was comparable for both groups). Notably such past evidence is based on parent-report assessment of sleep. The current study therefore aims to assess the extent to which these concurrent and longitudinal associations are mirrored in an objective measure of sleep.

The association between sleep and bedtime routines is an important but relatively understudied area in children with ASD. Bedtime routines are a component of sleep hygiene defined as “a set of observable, repetitive behaviours which directly involve the child and at least one adult acting in an interactive or supervisory role...in the hour preceding bed each night” (Henderson & Jordan, 2010, p. 272). The consistency of bedtime routines (e.g., consistent bed time, consistent sleep location) is associated with better parent-reported sleep quality in children with ASD (Henderson, Barry, Bader, & Jordan, 2011). In addition to the consistency of the routine, the activities present in the period preceding bedtime can also influence sleep quality. Specifically, the presence of ‘maladaptive activities’ (e.g., video games, active play, watching television, snacks/drinks) can increase physiological arousal and therefore delay sleep onset (Henderson & Jordan, 2010). Development across the school-age years is associated with more independence and a lessening of parental influence at bedtime. Consequently, increasing age is associated with a higher frequency of maladaptive activities in the hour before bedtime, in TD children (Henderson et al., 2011). Changes in the

quality of bedtime routines over time may therefore be expected to align with the course of sleep parameters over time, specifically those associated with the initiation of sleep.

The combination of both subjective and objective sleep measures is inherent to a comprehensive assessment of sleep. It has been proposed that a reliance on parent report for the assessment of sleep problems in children with ASD may be limited by the ‘overshadowing’ of sleep onset difficulties. Specifically, over identification of sleep problems may occur in those children who have difficulties in the initiation of sleep (Hodge, Parnell, Hoffman, & Sweeney, 2012). Consequently, multiple and objective methods of sleep assessment are recommended to increase the accuracy of sleep profiles. As asserted by Hodge et al. (2012), although polysmnography (PSG) is considered the ‘gold standard’ sleep assessment, its utility in ASD is limited. The use of sensors and an overnight laboratory stay are likely to trigger sensory sensitivities and negative reactions to the change in routine/environment. Where studies have employed PSG in ASD, sample sizes tend to be limited and the extent to which the data reflects the child’s typical sleep experience has been questioned (Hodge et al., 2012). Instead, actigraphy (an objective measure of sleep which infers sleep-wake patterns through the detection of movement) offers a non-invasive alternative which can be used in the home environment. In addition to traditional sleep parameters (e.g., sleep onset time, sleep duration, sleep efficiency), actigraphy also has the capacity to measure the stability of sleep; that being the extent to which sleep changes from night to night over a given period. Although there is some evidence that pre-school children (Anders, Iosif, Schwichtenberg, Tang, & Goodlin-Jones, 2011) and adults (Hare, Jones, & Evershed, 2006) with ASD are characterised by greater sleep variability than TD children, this has not yet been examined in school-aged children with ASD. Given the associations between sleep stability and daytime behaviour in TD children (Biggs, Lushington, van den

Heuvel, Martin, & Kennedy, 2011), characterisation of how the variability of sleep in ASD differs from TD children may provide novel insights into sleep in this paediatric population.

The current study therefore aimed to compare the trajectory of parent-report and actigraphy derived sleep profiles in a community recruited group of TD school-aged children and children with ASD, over a period of 12-15 months<sup>1</sup>. Furthermore, we aimed to examine the association between individual changes in sleep profiles and parent-reported anxiety and bedtime routines over this same period of time. It was therefore hypothesised that: i) changes in parent-reported sleep problems over the one year period would differ between groups (i.e., a significant interaction between group and time); ii) changes in objective sleep profiles over the same period of time would *not* differ between groups (i.e., a main effect of time only, for actigraphy derived sleep duration and time of sleep onset); iii) individual changes in parent-reported sleep problems over time would be associated with changes in anxiety and bedtime routines. Due to a lack of previous research utilising objective sleep measures, hypotheses were not made regarding the association between the actigraphy derived sleep-profiles and anxiety/bedtime routines.

## Method

### Participants

Participants were recruited from the XXXXXXXX project. Children in this sample were originally recruited through advertisements in local schools and community organisations. Children in the ASD group were additionally recruited through ASD support organisations, parent groups and advertisements in paediatric clinics throughout XXXXXX. All participants from the baseline sample who satisfied the following criteria were invited to take part in the follow-up study: i) parental consent provided at baseline to be contacted for

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<sup>1</sup> N.B. The sample in the current study is distinct and was recruited/tested subsequent to previous work from RC, NR, & KC (i.e., May et al., 2015).



future studies, ii) a full actigraphy dataset provided by the child at baseline, with participation occurring outside of school holidays, ii) a full scale IQ (FSIQ) >70 as measured by the Wechsler Abbreviated Scale of Intelligence (WASI; Wechsler, 1999), iii) verbal native English speaker (i.e., English as a primary language), iv) in the TD group only, children were additionally required to not have any siblings with ASD and to score below the clinical cut-off of the Social Responsiveness Scale (SRS; Constantino & Gruber, 2005).

Recruitment letters were therefore sent to seventy-seven participants, with follow up contact (i.e., phone call and/or e-mail) made by the first author within two weeks. Reasons for subsequent non-participation included the following: No response from e-mail/ phone call (ASD = 2, TD = 11), inconvenient timing cited by parents (ASD = 3, TD = 2), parental report at screening that child was unwilling to wear actiwatch at follow-up (ASD = 3), and the diagnosis of a health problem since baseline (1 child with ASD who was recently diagnosed with obstructive sleep apnoea). Consent was therefore acquired from 55 parents (23 parents of children with ASD and 32 parents of TD children). Return of participation materials resulted in the loss of 5 participants: 4 watches were returned unworn due to child illness or unforeseen circumstances (ASD = 2, TD = 2) and 1 watch experienced a malfunction and did not record any data (TD = 1). The final sample therefore consisted of fifty children (21 children with ASD and 29 TD children). Analyses were performed for each group to explore differences between children who took part in the follow up study, and those who did not. No significant differences were found for baseline age, CSHQ total score, or actigraphy derived sleep efficiency (all  $p > .05$ ).

All children within the ASD group had a diagnosis of Autistic Disorder or Asperger's Disorder in accordance with DSM-IV-TR (American Psychiatric Association, 2000) diagnostic criteria, made prior to the study. Upon entering the study, all parents provided a diagnostic report performed by either a paediatrician or registered psychologist and all reports

were checked against the DSM-IV-TR symptom checklist prior to participation. Parents in both groups completed the Social Responsiveness Scale (Constantino & Gruber, 2005) at baseline and follow-up, which was used to screen for ASD in the TD group (see above) and assist with the confirmation of diagnosis in the ASD group; whereby all children in the ASD group had a T score  $\geq 60$ .

Descriptive statistics for participant demographics and parent-report behavioural measures are shown in Table 1. Participant age ranged from 6 years, 2 months to 12 years 10 months at baseline, and 7 years, 2 months to 13 years 11 months at follow up. Across participant demographics, there were no significant differences between groups in age [baseline ( $p = .50$ ), follow-up ( $p = .50$ )], Ethnicity ( $p = .62$ ), SES score ( $p = .83$ ) or parental education ( $p = .089$ ). However, the TD group had a significantly higher Full Scale IQ (FSIQ) than the ASD group ( $p = .009$ ) and there was a significant association between group and gender ( $p = .019$ ). Regarding medication use, seven children in the ASD group were medicated at baseline (melatonin  $n = 3$ , stimulant  $n = 2$ , SSRI  $n = 2$ , atomoxetine  $n = 1$ ). Ten children were medicated at follow-up (melatonin  $n = 6$ , stimulant  $n = 6$ , SSRI  $n = 4$ ). No children had received any sleep treatment or sleep interventions between baseline and follow up.

[INSERT TABLE 1 HERE]

## Measures

### *Parent-report measures*

Sleep problems were assessed with the Children’s Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000), a 48 item caregiver-completed questionnaire with 8 subscales which align with the key sleep complaints relevant for school-aged children; bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night waking, parasomnias, sleep disordered breathing, and daytime sleepiness. Items are scored on a three-point likert scale from ‘rarely’ (1) to ‘usually’ (3), with higher scores indicative of greater sleep disturbance. The CSHQ demonstrates adequate internal consistency, acceptable test-retest reliability and discriminant validity. The clinical cut-off (total scores  $\geq 41$ ) identifies sleep disorders with a sensitivity of .80 and a specificity of .72 (Owens et al., 2000). CSHQ scores in the current sample had good internal consistency in the TD group (Cronbach’s  $\alpha = .89$ ) and acceptable internal consistency in the ASD group (Cronbach’s  $\alpha = .71$ ).

Anxiety symptoms were assessed with the Spence Children’s Anxiety Scale (SCAS; Spence, 1998), a 38 item caregiver-completed questionnaire. Responses are based on a four point likert scale from ‘never’ (0) to ‘always’ (3), with higher scores indicative of greater anxiety. The SCAS subscales coincide with the anxiety disorders listed in the DSM-IV-TR (American Psychiatric Association, 2000) and include: separation anxiety, physical injury fears, social phobia, obsessive compulsive, generalised anxiety disorder/overanxious disorder, and panic attack & agoraphobia. The reliability of the six SCAS subscales is high (Cronbach’s  $\alpha$  0.80 to 0.90) and convergent and discriminant validity have also been demonstrated (Nauta et al., 2004). SCAS scores in the current sample had good internal consistency in the both TD group and ASD group (Cronbach’s  $\alpha = .84$  & Cronbach’s  $\alpha = .89$ , respectively).

Parents also completed the Social Worries Questionnaire – Parent version (SWQ-P; Spence, 1995) which is a 10 item measure used to assess children’s fear and avoidance of social situation, from 0 ‘not true,’ to 2 ‘mostly true’. The scale has been used previously in

children with ASD (Russell & Sofronoff, 2005) and demonstrates high internal consistency (Cronbach's  $\alpha = .94$ ; Spence, 1995) and excellent criterion validity (Bailey, Chavira, Stein, & Stein, 2006). *SWQ scores in the current sample had acceptable internal consistency in the TD group (Cronbach's  $\alpha = .79$ ) and good internal consistency in the ASD group (Cronbach's  $\alpha = .83$ ).*

Bedtime routines were assessed with the Bedtime Routines Questionnaire (BRQ; Henderson & Jordan, 2010), a 31 item caregiver-report questionnaire with responses based on a 5 point likert scale from 'almost never' to 'nearly always.' Scores are provided for the consistency and reactivity of bedtime routines, along with the presence of adaptive and maladaptive activities in the hour before bedtime. The BRQ demonstrates acceptable to excellent internal consistency ( $\alpha = .69$  to  $.90$ ) and fair validity (Henderson & Jordan, 2010). *BRQ scores in the current sample had acceptable internal consistency in the both TD group and ASD group (Cronbach's  $\alpha = .77$  & Cronbach's  $\alpha = .75$ , respectively).*

### *Objective sleep profiles*

Participants were also provided with an Actiwatch-2 (Respironics Actiwatch 64, USA) to wear on their non-dominant wrist for fourteen consecutive nights, with each watch configured to record at 30 second epochs. Each actiwatch also features a marker button which was used by participants to indicate bedtime and get up time. Actigraphy data were analysed using the Respironics Actiware Software (Version 5.70.1) at medium sensitivity (40 activity counts per epoch required to be scored as 'wake'). The first epoch of ten consecutive minutes of immobility after bedtime was used to indicate time of sleep onset and the last epoch of ten consecutive minutes of immobility before get up time was used to indicate time of sleep offset. Sleep period duration was calculated as the number of minutes between sleep onset time and sleep offset. Sleep onset latency was calculated as the number of minutes from

bedtime to sleep onset time. Wake after sleep onset (WASO) was defined as the number of minutes scored as ‘wake’ during the sleep period. Sleep efficiency was calculated as the percentage of time spent asleep from time spent in bed (bedtime to get up time).

As recommended for the use of actigraphy in paediatric research, parents were provided with a sleep diary to complete across the two week period (Meltzer, Montgomery-Downs, Insana, & Walsh, 2012). Parents were asked to mark with an arrow their child’s bedtime and get up time, in addition to estimated time of sleep onset and offset. Completion of the sleep diary also required parents to note any daytime naps as well as any periods of time when the watch was removed from the wrist. Parents were also encouraged to document any issues which may have affected their child’s sleep (e.g., sleeping at a friend’s house, illness), and these nights of data were subsequently removed from analysis. All children were required to have at least 10 nights of actigraphy data. The sleep diary served to help eliminate artefact and to validate the marker/light out information provided from the actigraphy data.

**Procedure**

The study procedure was approved by XXXXXXXX Ethics Committee. Parental written consent and child assent was acquired for all participants. Participants subsequently received an actiwatch and relevant paperwork (consent form, health questionnaire, questionnaire booklet) via mail. Contact was made with all parents to confirm receipt of the actiwatch and to provide instructions on how to use the actiwatch and complete the sleep diary etc. Subsequent to the two-week recording period, all materials were returned via pre-paid mail. All children received 2 movie vouchers for their participation. All testing took place outside of school holidays, 12-15 months after baseline testing occurred. Upon study intake, all parents were asked to report any current health problems, the use of any prescription medication, and any sleep problems experienced since baseline assessment.

## Data Analyses

All parent-report and actigraphy variables were screened for outliers and any z-scores  $> \pm 3.29$  were recoded to 1 unit higher than the next non-outlier. This process resulted in the recoding of  $\leq 1$  value per variable. Associations between group and categorical outcome variables were assessed by Pearson chi-square or Fisher's exact test (where expected frequency assumptions were violated). Parent-report outcome measures (recorded at both baseline and follow-up) were analysed with a series of 2x2 mixed model ANOVAs to identify main effects of group and time, and significant interaction effects.

The CSHQ cut-off score of 41 was used to establish the presence of 'current' sleep problems at baseline and follow up. This was subsequently used to categorise participants as having persistent sleep problems ( $\geq 41$  at both time points), improved sleep problems ( $\geq 41$  at baseline only), acquired sleep problems ( $\geq 41$  at follow up only) or no sleep problems ( $< 41$  at both time-points).

For the actigraphy variables, both 'average' and 'variability' parameters are reported. The night-to-night variability of sleep was calculated as the average of the *absolute value* of the difference between each two consecutive nights of data (for example:  $| \text{day 1} - \text{day 2} | + | \text{day 2} - \text{day 3} | \dots \div \text{total number of nights}$ ). These variability parameters may subsequently be identified by a 'vari' suffix (e.g, SOTvari).

As performed recently in a similar study design (Andersen, Skogli, Hovik, Egeland, & Oie, 2015) difference scores were calculated for all outcome variables (i.e.,  $T2 - T1$ ), whereby a positive 'difference score' is indicative of an increase over time. Exploratory Pearson correlations were then performed across these difference scores to identify associations between change in sleep and behaviour over time. Fisher's r-to-z transformation

was used to compare the strength of these correlation coefficients between groups, whereby  $p > .05$  indicates the correlation coefficients are comparable.

Reported effect sizes include  $\eta_p^2$  (where values  $\geq .01 / \geq .06 / \geq .14$  = small/ medium/ large) and  $r^2$  (where values  $\geq .01 / \geq .09 / \geq .25$  = small/medium/large). Alpha was set at .05 for all analyses.

Results

The trajectory of sleep profiles over time

Given the significant group difference in gender and FSIQ, the association between these demographic variables and sleep were explored. FSIQ was not correlated with CSHQ total scores or any actigraphy derived sleep parameters (all  $p > .05$  and  $r^2 < .09$ ). In an examination of gender, TD males had a significantly lower ‘average’ sleep efficiency at baseline compared to TD females ( $p = .041$ ). No other significant associations between gender and sleep were identified.

Regarding parental-report, parents of children with ASD (61.9%) were significantly more likely to report that their child had experienced a sleep problem in the past year than parents of TD children (20.0%),  $\chi^2 = 9.28, p = .002$ . Additionally, there was a significant association between group and the persistence of CSHQ derived sleep problems,  $\chi^2 = 10.35, p = .010$ . Specifically, 76.2% of children with ASD had a ‘persistent’ sleep problem, compared to 30% of TD children. Conversely, 14.3% of children with ASD were free of current sleep problems at both time points, compared to 40% of TD children.

Descriptive statistics for the CSHQ are shown in Table 1. Children with ASD had significantly higher CSHQ total scores at both time points [ $F(1, 48) = 14.34, p < .001, \eta_p^2 = .23$ ]. Additionally, a significant interaction effect between group and time was observed for CSHQ Total Scores [ $F(1, 48) = 5.27, p = .031, \eta_p^2 = .10$ ], with more pronounced group



differences at baseline [ $F(1, 48) = 21.74, p < .001$ ] compared to follow-up [ $F(1, 48) = 5.66, p = .021$ ]. An examination of CSHQ subscales identified that this interaction was driven by significant group by time interaction effects in the bedtime resistance [ $F(1, 48) = 7.30, p = .001, \eta_p^2 = .13$ ] and sleep anxiety [ $F(1, 48) = 9.69, p = .003, \eta_p^2 = .17$ ] subscales. As previously noted, a small number of children within the ASD group commenced medication during the 12 month interval. Three of these children started taking melatonin to aid sleep during the 12 month period (one child started 8 months prior to T2, one child started 10 months prior to T2, and one child started 2 months prior to T2). It is therefore clarified that after the removal of these children, these interaction effects were still significant with a medium effect size: CSHQ sleep anxiety [ $F(1, 45) = 6.18, p = .017, \eta_p^2 = .12$ ] and CSHQ bedtime resistance [ $F(1, 45) = 4.55, p = .038, \eta_p^2 = .09$ ].

Table 2 provides descriptive statistics and the mixed model ANOVAs for the ‘average’ and ‘variability’ actigraphy parameters. Several main effects of group were identified between children with ASD and TD children. Across the ‘average’ sleep parameters, a significant main effect of group identified a lower average sleep efficiency in children with ASD than TD children ( $p = .003$ ). Given the effect of gender on sleep efficiency as detailed above, this mixed model ANOVA was repeated with gender added as a covariate. The significant main effect of group remained [ $F(1, 47) = 5.51, p = .024, \eta_p^2 = .10$ ]. The main effect of group for average sleep onset latency (SOL) approached significance, with a medium effect size ( $p = .063$ ). Across the ‘variability’ actigraphy parameters (e.g., SOT vari), significant main effects of group were identified for SOT vari ( $p = .045$ ), WASO vari ( $p = .029$ ), and SE vari ( $p = .002$ ); whereby children with ASD were characterised by greater night-to-night variability in sleep onset time, WASO, and sleep efficiency.

Several main effects of time were also identified, from baseline to follow-up (Table 2). Across the ‘average’ actigraphy parameters, significant main effects of time were



identified for sleep onset time ( $p = .001$ ) and sleep period duration ( $p < .001$ ), whereby increasing age was associated with a later sleep onset time and shorter sleep period duration. Across the ‘variability’ actigraphy parameters (e.g., SOT vari), significant main effects of time were identified for SOT vari ( $p = .008$ ), SOffT vari ( $p = .003$ ) and SD vari ( $p = .010$ ); whereby night-to-night variability in sleep onset time, sleep offset time, and sleep period duration was greater at follow-up, compared to baseline. No significant interaction effects were observed between group and time for any actigraphy parameter (all  $p > .05$ ), indicating the developmental trajectory of sleep profiles was comparable between groups.

[INSERT TABLE 2 HERE]

**Developmental associations between sleep, anxiety, and bedtime routines**

Descriptive statistics for parent-report measures of anxiety and bedtime routines are shown in Table 1. Several significant main effects of group were identified. Children with ASD were characterised by higher anxiety across both the SCAS [ $F(1, 48) = 15.80, p < .001, \eta_p^2 = .25$ ], and SWQ [ $F(1, 48) = 7.80, p = .008, \eta_p^2 = .14$ ]. Regarding bedtime routines, children with ASD were also characterised by more reactive bedtime routines [ $F(1, 48) = 20.50, p < .001, \eta_p^2 = .30$ ], more frequent maladaptive activities [ $F(1, 48) = 10.73, p = .002, \eta_p^2 = .18$ ] and less frequent adaptive activities [ $F(1, 48) = 4.60, p = .037, \eta_p^2 = .09$ ], in the hour before bedtime. A significant main effect of time on BRQ consistency scores indicated a reduction in the consistency of bedtime routines at follow up, compared to baseline [ $F(1, 48) = 5.55, p = .023, \eta_p^2 = .10$ ]. No significant interaction effects were identified across these

measures, indicating the change in these variables over time was comparable between children with and without ASD.

To demonstrate the large amount of heterogeneity in individual change over the one year period, descriptive statistics for difference scores are shown in Table 3. Mirroring the significant interaction effect (group x time), there was a significant difference in CSHQ total difference scores [ $t(48) = 2.18, p = .034$ ], indicating a greater reduction in parent-reported sleep problems in children with ASD than TD children. Average difference scores for all other variables were comparable between groups (all  $p > .05$ ). Age was not associated with difference scores in CSHQ, sleep efficiency, or sleep period duration (all  $p > .05$  and  $r^2 < .09$ ).

[INSERT TABLE 3 HERE]

Pearson correlations between changes in sleep, anxiety, and bedtime routines are shown in Table 4. Regarding parent-reported sleep problems, change in anxiety was positively correlated with change in CSHQ total scores, across both SCAS total scores [ $(r = .58, p < .001)$ ; (Fisher's r-to-z transformation,  $Z = .17, p = .34$ )] and SWQ total scores [ $(r = .32, p = .027)$ ; (Fisher's r-to-z transformation,  $Z = 1.42, p = .08$ )]. The relationship between change in CSHQ total scores and change in BRQ reactivity scores approached significance ( $r = .27, p = .054$ ).

Regarding actigraphy derived sleep profiles, change in the frequency of maladaptive behaviours in the hour before bedtime was negatively correlated with change in 'average' sleep efficiency [ $(r = -.38, p = .006)$ ; (Fisher's r-to-z transformation,  $Z = -.39, p = .70$ )]. That is, children who increased the frequency of maladaptive behaviours over time experienced a

reduction in sleep efficiency, and vice versa. Change in the frequency of maladaptive behaviours in the hour before bedtime was also positively correlated with changes in the night-to-night variability of sleep efficiency (i.e., SE vari;  $r = .33, p = .021$ ). However, this relationship did not hold after controlling for average sleep efficiency ( $r = .13, p = .37$ ). As shown in Table 4, the relationship between sleep efficiency and maladaptive behaviours appeared to be driven by the positive correlation between change in the frequency of maladaptive behaviours and change in sleep onset latency [ $(r = .30, p = .037)$ ; (Fisher's r-to-z transformation,  $Z = .48, p = .63$ )]. That is, children who increased the frequency of maladaptive behaviours over time experienced an increase in the time taken to initiate sleep (SOL). These significant associations held after the removal of children who commenced medication during participation: maladaptive activities and sleep efficiency difference scores,  $r = -.40, p = .005$ ; and maladaptive activities and sleep efficiency difference scores,  $r = -.32, p = .029$ . Given that sleep efficiency is a composite measure accounting for night wakings and sleep onset latency, it is also noted that change in sleep efficiency was correlated with change in SOL [ $r = -.77, p < .001$ ], but not WASO [ $r = -.15, p = .31$ ]. As shown in Table 5, a significant correlation was also identified between change in social anxiety (i.e., SWQ scores) and change in sleep onset latency ( $r = .32, p = .042$ ). It is noted however, that this correlation did not hold after controlling for chronological age [ $r(47) = -.27, p = .065$ ].

[INSERT TABLE 4 HERE]

## Discussion

The current study assessed the trajectory of parent-report and objective sleep profiles over a one year period in school-aged children with and without ASD. As hypothesised, the change in parent-reported sleep problems over time was dependent on group, with a significant interaction between group and time for CSHQ total scores. In line with hypothesis two, comparable trajectories were identified across groups in objective sleep profiles. Both children with ASD and TD children had a significantly later sleep onset time and shorter sleep duration at follow-up compared to baseline. Additionally, both groups were characterised by a significant increase in the night-to-night variability of sleep onset time and sleep period duration at follow-up compared to baseline. Hypothesis three was partly supported change in CSHQ total scores were associated with changes in symptoms of anxiety, but not bedtime routines. Given the exploratory nature of this research, hypotheses were not made regarding the association between actigraphy, sleep and bedtime routines. This analysis identified change in maladaptive behaviours were positively associated with change in sleep efficiency, specifically the time take to initiate sleep (SOL).

In line with cross-sectional evidence (Hodge et al., 2014) and more recent longitudinal findings (May et al., 2015), improvements in parent-reported sleep problems over a one year period were evident only in the ASD group, characterised by reductions in sleep anxiety and bedtime resistance. This reinforces the notion that behavioural sleep problems may be at their most pronounced during early childhood for children with ASD (Hodge et al., 2014; Wiggs & Stores, 2004). One previous study has assessed the trajectory of objective sleep profiles in school-aged children with ASD relative to TD children, with this study conducting a 2-3 year follow up (Allik et al., 2008). In line with their findings, there were no significant interactions between group and time (i.e., the trajectory was similar

across groups), with a significant decrease in sleep duration across both groups, and no change over time in sleep onset latency. Discrepant with their findings however, the current study did not identify a reduction in sleep efficiency, or an increase in night waking. This may reflect the shorter follow-up time used in the current study.

The increased night-to-night variability in the ASD group complements previous findings in pre-school children (Anders et al., 2011) and adults (Hare et al., 2006) with ASD. Furthermore, an increase in the variability of sleep was also seen across both groups at follow-up assessment. However, the clinical implications of sleep variability are poorly understood across all study populations, due to ‘ad-hoc’ examinations and inconsistent methodological approaches in previous research (Bei, Wiley, Trinder, & Manber, 2016). Nonetheless, future research should explore which dynamic factors may influence the increased variability of sleep in ASD (e.g., diet, sleep hygiene, anxiety).

Regarding the examination of difference scores, individual change in anxiety and social worries was positively associated with change in the severity of parent-reported sleep problems. This complements past research identifying significant concurrent associations between parent-reported sleep and anxiety (Goldman et al., 2011; Hollway et al., 2013; Mayes & Calhoun, 2009; Rzepecka et al., 2011). **Future research may therefore examine whether** elevated levels of anxiety in young children with ASD exacerbate sleep problems during a developmental period which is already vulnerable to a variety of sleep related problems. Notably the relationship between change in anxiety and change in parent-reported sleep problems was comparable between groups. This supports the notion that although sleep problems are more severe in children with ASD, non-clinical anxiety symptoms may be associated with parent-reported sleep concerns in typical, as well as atypical, development (Fletcher et al., in press). However, there was no association between anxiety and actigraphy derived sleep profiles. This may be interpreted in two ways. Firstly, this may highlight the

limitations of correlating two parent-report measures, whereby associations may be inflated. In the context of the CSHQ and actigraphy however, this may suggest that anxiety has a particular association with sleep behaviours which both precede and follow the sleep period (e.g., sleep related anxiety, daytime sleepiness), and are therefore not directly measurable by actigraphy. This again reinforces the need to use both subjective and objective measures of sleep.

There was substantial variability in sleep efficiency ‘difference scores’ across both groups (Table 3). Such heterogeneity suggests large individual differences in the maturation of sleep during this time. Of clinical importance therefore, is the need for future studies to clarify which factors predict an improvement or worsening of sleep quality. The current findings suggest that maladaptive pre-bedtime behaviours may play a key role. An association was identified between an increase in the frequency of maladaptive activities in the hour before bedtime and decreased sleep efficiency, impacting on sleep onset latency. Previous research identifying an association between bedtime routines and actigraphy derived sleep profiles in TD children questioned whether bedtime routines are a direct mechanism for stress reduction (and therefore better sleep quality), or whether they serve as a mediator between other variables such as parent behaviour management, household environment, or child temperament (Mindell, Li, Sadeh, Kwon, & Goh, 2015). In terms of maladaptive behaviours however, playing video games before bedtime has been shown to increase heart rate and significantly increase the time take to initiate sleep (Higuchi, Motohashi, Liu, & Maeda, 2005). Given that children in the ASD group were characterised by increased maladaptive activities across both time points, this association requires further exploration in future studies. Actigraphy monitoring alongside a diary of pre-bedtime activities would serve to validate this link and assess the direct association between activities in the hour before bedtime and the subsequent nightly sleep quality.

The current study is limited by the relatively small sample size and subsequently reduced statistical power. It is noted however, that most medium effect sizes across the analyses reached statistical significance (the exceptions being the main effects of group for average SOL, SOL vari, and SD vari, in addition to the interaction effects for the SOffT vari and WASO vari). Additionally, a measure of pubertal stage was not used and it was therefore not possible to ascertain the effect of pubertal development on difference scores over the one year period.

The current study support previous findings that children with ASD are characterised by a lower sleep efficiency than their TD peers, across the school-age years. Conversely, parents of children with ASD report a reduction in pre-sleep behavioural sleep problems over this same period, reducing the magnitude of group differences. A large amount of variability was seen in the difference scores for behavioural and objective measures, with those children who experienced a reduction in sleep quality characterised by an increase in anxiety symptoms and an increased frequency of maladaptive activities in the hour before bedtime. Future research should seek to track daily anxiety and maladaptive pre-sleep behaviours alongside actigraphy monitoring, in order to map the dynamic changes between these factors.

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Table 1

*Participant characteristics, presented as M (SD) or n (%)*

|                             | Baseline       |                 | Follow-up      |                 |
|-----------------------------|----------------|-----------------|----------------|-----------------|
|                             | TD<br>(n = 29) | ASD<br>(n = 21) | TD<br>(n = 29) | ASD<br>(n = 21) |
| Age, months                 | 102.10 (17.07) | 106.67 (26.82)  | 115.97 (16.81) | 120.52 (29.94)  |
| Gender, n male              | 14 (48.3)      | 17 (81.0)       | -              | -               |
| Full Scale IQ               | 111.97 (13.21) | 101.29 (13.92)  | -              | -               |
| Ethnicity                   |                |                 |                |                 |
| Caucasian                   | 27 (93.1)      | 20 (95.2)       | -              | -               |
| Other                       | 2 (6.9)        | 1 (4.8)         | -              | -               |
| SES Score                   | 7.41 (2.64)    | 7.57 (2.42)     | -              | -               |
| Primary caregiver education |                |                 |                |                 |
| Postgraduate                | 4 (13.8)       | 2 (9.5)         | -              | -               |
| Undergraduate               | 15 (51.7)      | 17 (81.0)       | -              | -               |
| High School                 | 10 (34.5)      | 2 (9.5)         | -              | -               |
| CSHQ total score            | 40.87 (4.96)   | 50.14 (9.02)    | 42.90 (8.58)   | 48.19 (6.46)    |
| SCAS total score            | 12.76 (8.36)   | 24.24 (10.60)   | 14.21 (8.87)   | 23.67 (13.25)   |
| SWQ total score             | 3.90 (3.63)    | 7.19 (4.27)     | 3.97 (3.35)    | 6.55 (4.52)     |
| BRQ                         |                |                 |                |                 |
| Reactivity                  | 7.55 (2.64)    | 11.10 (4.84)    | 6.49 (2.19)    | 10.86 (4.49)    |
| Consistency                 | 42.31 (6.38)   | 42.61 (4.62)    | 40.03 (6.58)   | 41.10 (7.56)    |
| Maladaptive Activities      | 11.55 (3.38)   | 14.67 (3.23)    | 11.59 (4.25)   | 14.48 (3.61)    |
| Adaptive Activities         | 43.03 (4.30)   | 40.05 (5.45)    | 42.79 (4.72)   | 40.52 (4.88)    |

*Note.* ASD: Autism Spectrum Disorder; BRQ: Bedtime Routines Questionnaire; CSHQ: Children's Sleep Habits Questionnaire; SCAS: Spence Children's Anxiety Scale; SES: Socio-Economic Status; SWQ: Social Worries Questionnaire; TD: Typically Developing.

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Table 2

2 x 2 Mixed Model ANOVAs for main effects of group (ASD & TD), time (Baseline to follow-up), and interaction effects for actigraphy variables.

|               | TD Group<br>(n = 29) |                | ASD Group<br>(n = 21) |                | Group   |            | Time     |            | Interaction |            |
|---------------|----------------------|----------------|-----------------------|----------------|---------|------------|----------|------------|-------------|------------|
|               | Baseline             | Follow-up      | Baseline              | Follow-up      | F       | $\eta^2_p$ | F        | $\eta^2_p$ | F           | $\eta^2_p$ |
| SOT (hh:mm)   | 21:23 (0:41)         | 21:36 (0:41)   | 21:20 (0:39)          | 21:39 (0:41)   | <.01    | <.01       | 11.78*** | .20        | 0.40        | .01        |
| SOffT (hh:mm) | 6:54 (0:32)          | 6:56 (0:34)    | 6:51 (0:30)           | 6:46 (0:32)    | 0.53    | .01        | 0.12     | <.01       | 0.54        | .01        |
| SD (min)      | 571.10 (31.19)       | 559.24 (23.43) | 564.86 (35.19)        | 547.17 (33.33) | 1.13    | .02        | 21.19*** | .31        | 0.82        | .02        |
| SOL (min)     | 23.70 (12.28)        | 25.87 (12.33)  | 32.30 (20.23)         | 32.76 (20.28)  | 3.62    | .07        | 0.37     | <.01       | 0.16        | <.01       |
| WASO (min)    | 55.39 (12.11)        | 53.98 (12.21)  | 57.61 (18.14)         | 53.52 (17.06)  | 0.04    | <.01       | 3.65     | .07        | 0.87        | .02        |
| SE (%)        | 91.11 (2.74)         | 90.97 (2.09)   | 89.08 (3.88)          | 88.45 (3.76)   | 9.57**  | .17        | 0.61     | .01        | 0.25        | 0.01       |
| SOT vari      | 0:33 (0:12)          | 0:39 (0:15)    | 0:32 (0:15)           | 0:41 (0:29)    | 0.01    | <.01       | 7.76**   | .14        | 0.16        | <.01       |
| SOffT vari    | 0:33 (0:10)          | 0:37 (0:14)    | 0:34 (0:09)           | 0:47 (0:17)    | 4.23*   | .08        | 9.80**   | .17        | 3.18        | .06        |
| SD vari       | 47.36 (13.12)        | 51.69 (16.39)  | 50.08 (17.65)         | 65.13 (32.98)  | 3.02    | .06        | 7.29**   | .13        | 2.23        | .04        |
| SOL vari      | 13.63 (6.70)         | 18.62 (11.50)  | 20.67 (12.88)         | 20.88 (15.33)  | 2.82    | .06        | 1.94     | .04        | 1.65        | .03        |
| WASO vari     | 13.67 (6.06)         | 14.94 (6.25)   | 19.30 (8.20)          | 16.33 (7.19)   | 5.04*   | .10        | 0.51     | .01        | 3.18        | .06        |
| SE vari       | 4.42 (1.88)          | 5.94 (1.76)    | 5.64 (2.02)           | 4.94 (1.76)    | 10.50** | .18        | 2.75     | .05        | 0.14        | <.01       |

Note. ASD: Autism Spectrum Disorder; SD: sleep **period** duration; SE: sleep efficiency SOffT: sleep offset time; SOL: sleep onset latency; SOT: sleep onset time; TD: typically developing; ‘Vari’ suffix: night-night-variability; WASO: wake after sleep onset; \* $p \leq .05$ , \*\* $p \leq .01$ , \*\*\* $p \leq .001$ .

Table 3

*Mean (M), Standard Deviations (SD) and range of difference scores across measures for the TD and ASD groups*

|                 | TD ( <i>n</i> = 29) |        |       | ASD ( <i>n</i> = 21) |        |       |
|-----------------|---------------------|--------|-------|----------------------|--------|-------|
|                 | <i>M (SD)</i>       | Min    | Max   | <i>M (SD)</i>        | Min    | Max   |
| Actigraphy      |                     |        |       |                      |        |       |
| SD (min)        | -11.86 (19.34)      | -55.23 | 34.43 | -17.68 (26.09)       | -59.24 | 43.11 |
| SOL             | 2.17 (11.71)        | -31.37 | 35.93 | -0.13 (17.34)        | -25.75 | 39.51 |
| WASO            | -1.41 (7.66)        | -20.54 | 14.43 | -4.08 (12.63)        | -25.64 | 18.57 |
| SE (%)          | -0.13 (3.11)        | -6.32  | 6.42  | -0.63 (3.84)         | -7.99  | 6.28  |
| Parent-report   |                     |        |       |                      |        |       |
| CSHQ Total      | 1.72 (5.90)         | -7.00  | 21.00 | -1.95 (5.87)         | -15.00 | 6.00  |
| SCAS Total      | 1.44 (9.06)         | -15.00 | 29.00 | 0.57 (8.08)          | -10.00 | 20.00 |
| SWQ Total       | 0.07 (3.32)         | -9.00  | 6.00  | -0.45 (3.49)         | -9.00  | 7.00  |
| BRQ Mal         | 0.04 (3.48)         | -5.00  | 9.00  | 0.00 (3.28)          | -7.00  | 6.00  |
| BRQ Adaptive    | -0.24 (3.66)        | -14.00 | 5.00  | 5.16 (0.48)          | -16.00 | 11.00 |
| BRQ Consistency | -2.28 (6.04)        | -17.00 | 12.00 | -1.52 (4.99)         | -13.00 | 6.00  |
| BRQ Reactivity  | 1.00 (3.00)         | -8.00  | 7.00  | -0.24 (4.32)         | -10.00 | 8.00  |

*Note.* ASD: Autism Spectrum Disorder; BRQ: Bedtime Routines Questionnaire; CSHQ: Children's Sleep Habits Questionnaire; SCAS: Spence Children's Anxiety Scale; SD: sleep period duration; SE: sleep efficiency; SWQ: Social Worries Questionnaire; TD: Typically Developing



Table 4

*Pearson bivariate correlations between difference scores across sleep and behavioural measures (N = 50)*

|                  | BRQ<br>Consistency | BRQ<br>Reactivity | BRQ<br>Adaptive | BRQ<br>Maladaptive | SCAS<br>Total | SWQ<br>Total |
|------------------|--------------------|-------------------|-----------------|--------------------|---------------|--------------|
| CSHQ total score | .10                | .27               | -.11            | .17                | .58***        | .32*         |
| SD (min)         | .01                | .01               | -.21            | -.14               | .13           | .02          |
| SOL (min)        | -.14               | -.16              | .01             | .30*               | -.17          | -.29*        |
| WASO (min)       | -.10               | .17               | -.01            | .20                | .14           | .09          |
| SE (%)           | -.02               | -.11              | -.02            | -.38**             | -.05          | .19          |

*Note.* BRQ: Bedtime Routines Questionnaire; CSHQ: Children’s Sleep Habits Questionnaire; SCAS: Spence Children’s Anxiety Scale; SD: sleep period duration; SE: sleep efficiency; SWQ: Social Worries Questionnaire; \*  $p \leq .05$ , \*\*  $p \leq .01$ . \*\*\*  $p \leq .001$ .